Open Journal of Stomatology, 2013, 3, 347-353 **OJST**
http://dx.doi.org/10.4236/ojst.2013.37059 Published Online October 2013 (http://www.scirp.org/journal/ojst/)

Hybrid ameloblastoma in a nigerian: Report of a case and review of literature

O. A. Effiom[1*], O. James[2], O. T. Akeju[3], A. S. Salami[3], O. Odukoya[1]

[1]Department of Oral Pathology and Oral Biology, College of Medicine, University of Lagos, Lagos University Teaching Hospital, Lagos, Nigeria
[2]Department of Oral and Maxillofacial Surgery, College of Medicine, University of Lagos, Lagos, Nigeria
[3]Department of Oral Pathology and Oral Biology, Lagos University Teaching Hospital, Lagos, Nigeria
Email: [*]jumokeffiom@yahoo.com

Received 8 July 2013; revised 8 August 2013; accepted 8 September 2013

ABSTRACT

Background: Hybrid Ameloblastoma is a rare type of ameloblastoma that is presently receiving attention in some quarters. Altogether, less than 30 cases have been reported in the scientific literature, thus calling for the need to report more cases in order to add to the body of knowledge on this lesion. This case report is aimed to add to the number of cases that are building up in the scientific literature. Materials and Methods: A 50-year-old female presented with a 3-year history of an anterior mandibular swelling extending from the body of the mandible on the right to the body of the mandible on the left. Orthopantomograph of the lesion revealed a mixed radiolucent and radiopaque lesion that extended from the distal region of the right mandibular second premolar to distal region of the left mandibular second molar. Segmental mandibulectomy with immediate replacement with reconstruction was undertaken. Results: Specimen which was subjected to histopathologic examination reported a diagnosis of Keratinizing Follicular Ameloblastoma and Desmoplastic Ameloblastoma with Osteoplasia, consistent with a diagnosis of Hybrid Ameloblastoma. There has been no sign of recurrence 7 months after a regular follow-up and the patient is planned for a long term follow-up. Conclusion: Besides histological details that combine both conventional ameloblastoma and desmoplastic ameloblastoma, the present case of hybrid ameloblastoma bears similar clinical features as well as treatment modalities to that of conventional solid multicystic ameloblastoma.

Keywords: Ameloblastoma; Hybrid; Nigerian

[*]Corresponding author.

1. INTRODUCTION

Ameloblastoma is a benign odontogenic tumour of epithelial origin [1,2]. It is found exclusively within the maxillofacial skeleton (intraosseous or centrally located) but has been reported to be located in the soft tissue (gingiva) overlying tooth bearing areas or alveolar mucosa in edentulous regions (extraosseous or peripherally located) [2]. The WHO in 2005 [2], categorized the conventional ameloblastoma into 4 clinical types namely: solid multicystic, unicystic, peripheral and desmoplastic (DA) clinical types. Ameloblastoma is the most common odontogenic tumour [3], but its precise etiology till date remains unknown although various etiologies have been postulated in the scientific literature [4,5]. It is the most commonly reported tumour in underdeveloped countries [6] with its highest prevalence recorded in Asian and African countries [7]. It has been reported to account for 73.0% of odontogenic tumours and 24.0% of tumours and tumour-like lesions of the jaws [8] and has been observed to occur in both sexes with an equal sex distribution [9] although a slight male predominance was been reported [10]. Ameloblastoma affects all age groups with an average age of 31.67 years but has a site predilection for the posterior mandible [11]. It usually presents clinically as an asymptomatic jaw swelling.

The main histologic types of the conventional ameloblastoma include the follicular and plexiform types although types that cannot be qualified as such are categorized as mixed types [12]. DA is an unusual variant of the conventional ameloblastoma. It presents with specific radio histologic features and anatomic distribution that make it different from other conventional types [13]. Histologically it is composed of irregular bizarre shaped proliferating islands and cords of odontogenic epithelium of different sizes that are embedded in a highly col-

laginized connective tissue stroma. Waldron and El-Mofty [14] discovered a rare variant of ameloblastoma composed histologically of areas of the classic follicular or plexiform ameloblastoma and areas of DA. This was named hybid ameloblastoma (HA).

Ameloblastoma commonly presents radiologically as a unilocular or multilocular lesion [15] although DA with osteoplasia and some cases of HA may present with the more unusual mixed radiolucent and radiopaque pattern similar to the radiological presentation of fibro-osseous lesions [15,16]. This uncommon radiological presentation of ameloblastoma may in some cases pose a diagnostic challenge particularly to inexperienced pathologists and surgeons, and may in such cases result in treatment delay. It is therefore important that the clinical, radiological and histological presentations of ameloblastoma is well recognized to prevent histological misdiagnosis among its various types and to differentiate ameloblastoma from other jaw lesions (e.g. fibro osseous lesions) which may have similar clinic-radiologic presentations, thus preventing histological misdiagnosis and inappropriate patient management. Perusal of the scientific English literature reveals that less than 30 cases of HA have been reported [17]. Presently we present a case of HA, a rare type of ameloblastoma and add this to the scientific literature. In Nigeria, only 2 cases have been reported [18]. As far as we know, our case would be the 3rd documented Nigerian HA reported so far.

2. CASE REPORT

A 50 year old woman presented in the Oral and Maxillofacial clinic of the Lagos University Teaching Hospital, with a well circumscribed anterior mandibular swelling of 3 years duration (**Figure 1**). Clinical examination revealed a tumour mass which measured 15 cm × 13 cm and extended from the mandibular premolar region on the right to the mandibular 1st molar region on the left (**Figure 2**). The tumour was firm to fluctuant in consistency, caused buccal and lingual expansion of the mandible and displaced the anterior mandibular teeth. An orthopantomograph (OPG) was taken and radiographically the tumour appeared as a mixed radiolucent and radiopaque lesion with irregular borders primarily located in the anterior mandible. The mandibular cortical bone expansion and displacement of the lower anterior teeth which were clinically observed were confirmed by the radiograph (**Figure 3**). Clinical impressions of ameloblastoma and ossifying fibroma were made based on the history and clinic radiological presentations. With an informed consent, an incisional biopsy was performed and a diagnosis of follicular acanthomatous ameloblastoma was reported (**Figure 4**). Based on the histological diagnosis under general anaesthesia, a segemental resection was performed followed by an immediate reconstruction of the defect in January 2013.

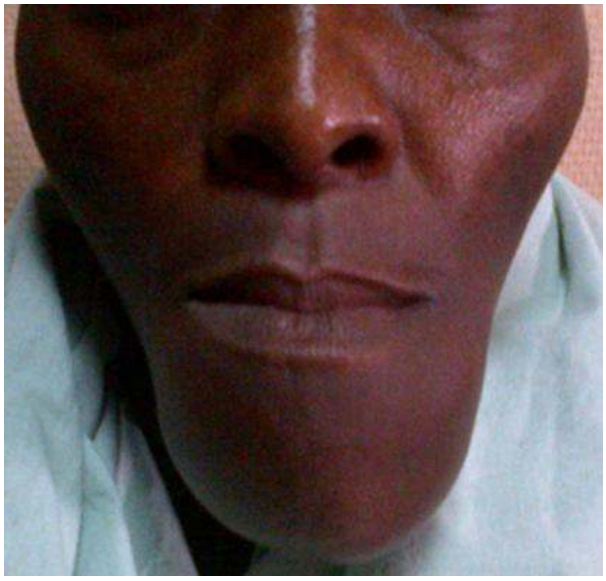

Figure 1. Clinical photograph showing gross anterior mandibular swelling.

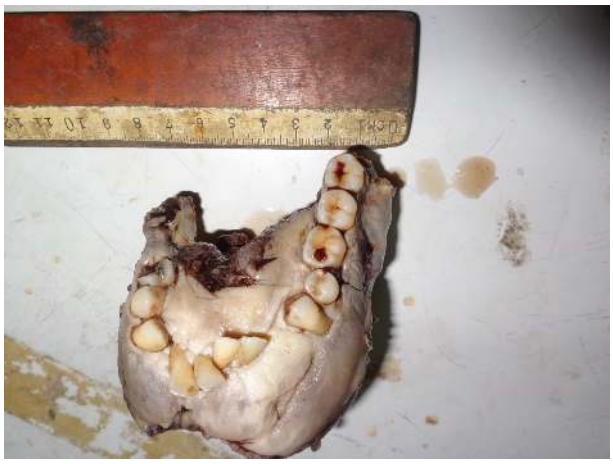

Figure 2. Resected mandible showing well circumscribed lesion with buccal and lingual cortical plate expansion and displacement of anterior teeth.

The post-surgical specimen was subjected to histological evaluation and this revealed the presence of ameloblastoma composed of different histological types. In an area, connective tissue stroma contained odontogenic epithelial cells arranged in follicles of peripheral columnar cells bordering loose central stellate reticulum-like cells that presented with squamous metaplasia, keratin pearl formation and cystic degeneration (conventional ameloblastoma area) (**Figure 5**). In another area, dense connective tissue stroma containing small islands of compressed odontogenic epithelium was observed. The compression of the follicles resulted in various figurative pattern presentation of the odontogenic epithelial

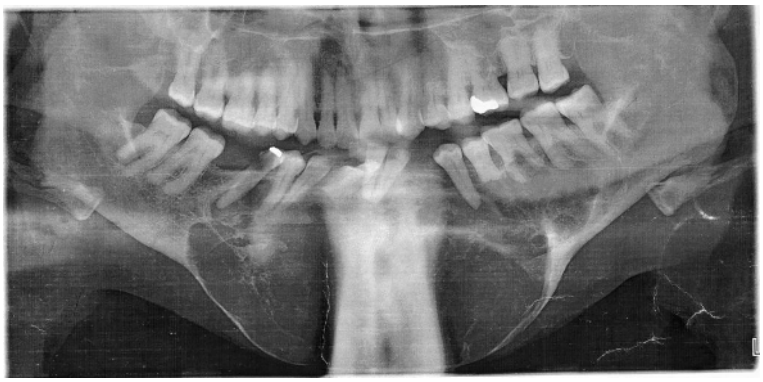

Figure 3. OPG showing marked cortical expansion and thinning of cortical bone, displacement of anterior mandibular teeth, a well defined irregular multilocular radiolucency and radiopaque specks within the radiolucent area resulting in a mixed radioluency and radioopacity.

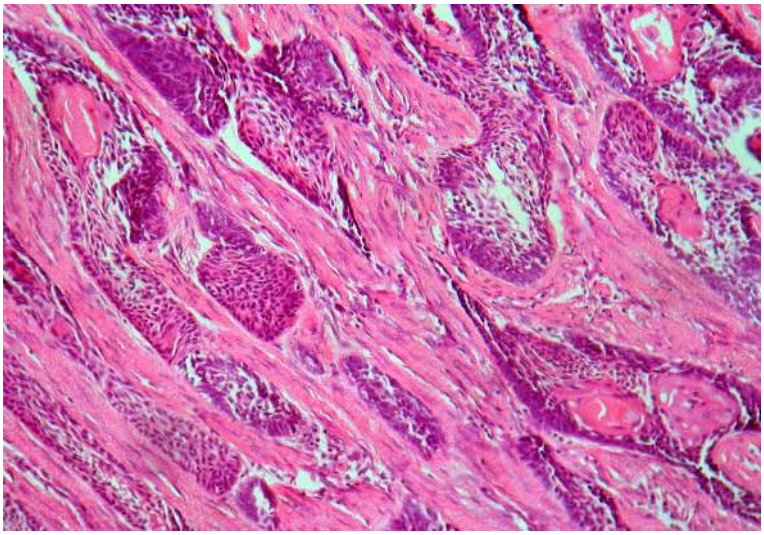

Figure 4. Islands of ameloblastomatous follicles in a connective tissue stroma. Central stellate reticulum cells showing squamous metaplasia in areas, H & E ×40.

cells (**Figure 6**). Areas of osseous metaplasia (osteoplasia) were also observed within the dense connective tissue stroma (desmoplastic ameloblastoma with osteoplasia area) (**Figure 7**). A definitive diagnosis of Keratinizing Follicular Ameloblastoma and DA with Osteoplasia, consistent with HA was made.

The patient's post-operative orthopanthomogram and clinical follow-up after 7 months disclosed no sign of recurrence and the patient is planned for a long term follow-up (**Figure 8**).

3. DISCUSSION

DA is an uncommon benign locally infiltrative clinico-surgical type of ameloblastoma. It has been reported to have incidence rates that range from 4.0% - 13.0% [15,16,19]. Histologically, it consists of irregular bizarrely shaped proliferating islands and cords of odontogenic epithelium of different sizes which are embedded in a highly collaginized connective tissue stroma (desmoplasia) [15,16,19]. HA on the other hand is a rare type of ameloblastoma that combines histologic features of both DA and conventional ameloblastoma. It has been reported to account for between 1.1% to 4.3% of ameloblastoma [13,17,18,20,21] and was initially discovered by Waldron and El-Mofty in 1987 to have a predilection for the posterior aspect of the mandible [14]. Presently we report one case of HA primarily located in the anterior mandible but with a posterior extension. This sole case being the only case documented and reported in 40 years, therefore accounts for 0.003% of the series of ameloblastoma reported at the Lagos University Teaching Hospital (LUTH). Predominant anterior posterior mandibular tumour site locations of HA have been observed in an earlier study of HA by Takata *et al.* [22]

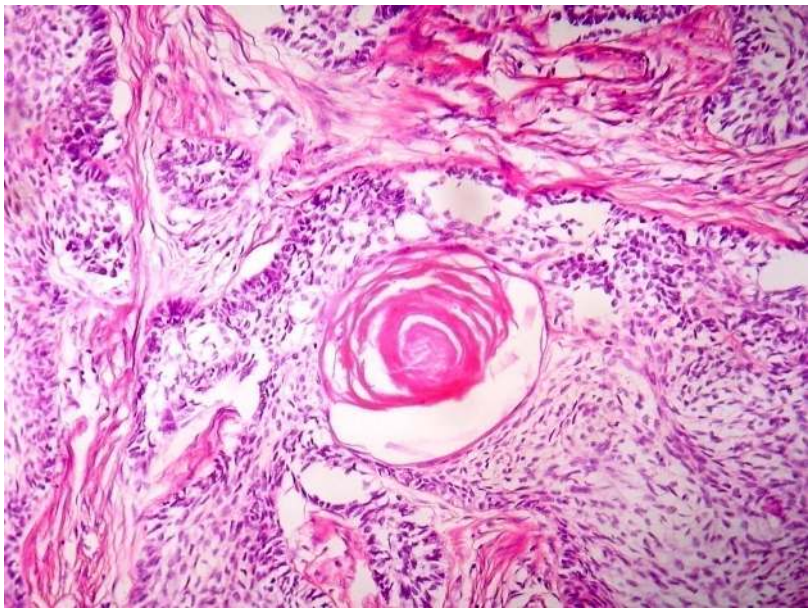

Figure 5. Islands of odontogenic epithelial cells with peripheral tall columnar cells and loose central stellate reticulum-like cells showing areas of squamous metaplasia and keratin pearl formation, H & E ×40.

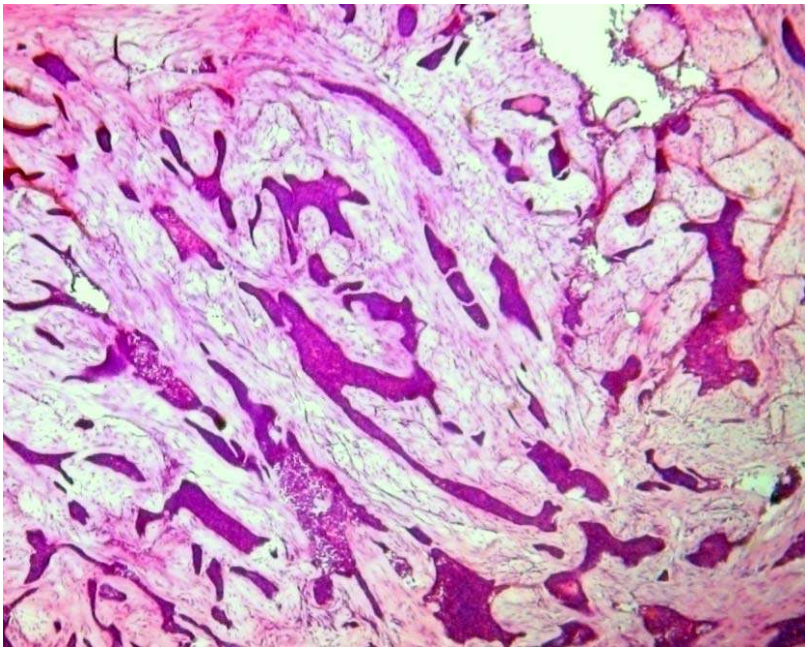

Figure 6. Islands of odontogenic neoplastic epithelial cells in various patterns supported by a dense connective tissue stroma, H & E ×40.

while a sole anterior mandibular predilection was reported in a previous Nigerian study [18].

Reports from the scientific literature show that most cases of HA present as mixed radiolucent and radiopaque lesions with irregular borders similar to the common radiological pattern observed in DA (with osteoplasia) or fibro osseous lesions although a few cases of HA have been observed to present radio graphically as multi locular radiolucencies similar to the common radiographic pattern of conventional ameloblastoma [15-18]. There has been suggestions that the radiographic presentation of HA (irregular ill-defined borders and mixed radiolucency and radioopacity) may express an infiltrative nature implying HA to be a more aggressive lesion than other variants of ameloblastoma [13,22]. Although this case presented radiographically as a mixed radiolucent

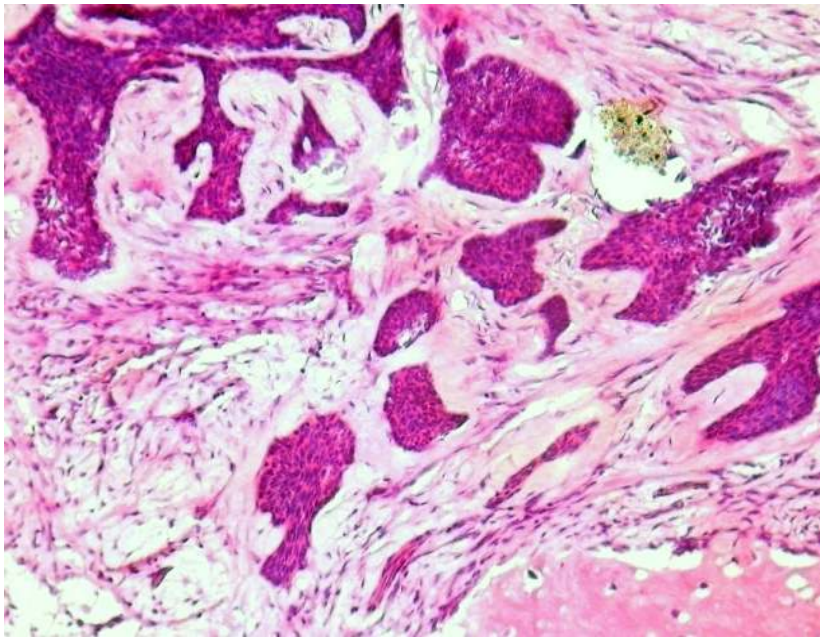

Figure 7. Area of osteoplasia within the collagenized fibrous connective tissue stroma in HA, H & E ×40.

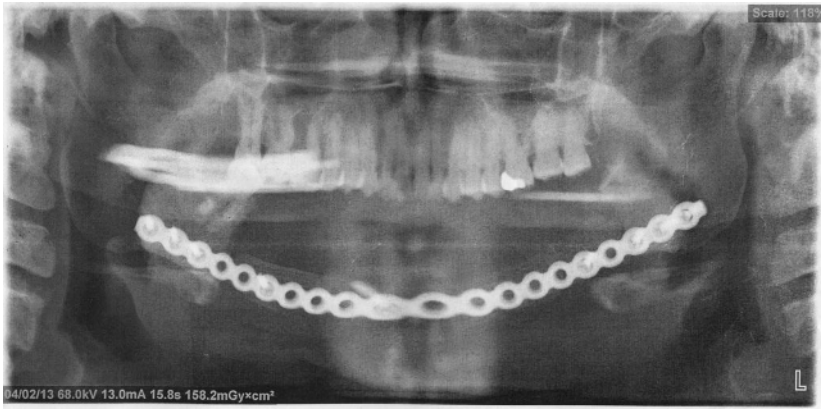

Figure 8. Post-operative radiograph of the patient after 7 months disclosed absence of lesion.

and radiopaque lesion with irregular well defined borders, it however still presented with similar clinical as well as similar treatment modalities to that of the conventional solid multicystic ameloblastoma. The presence of osseous metaplasia (osteoplasia) observed within the desmoplastic areas of the lesion may be the result of the peculiar radiographic presentation of the lesion. It is worth nothing that HA has been observed to share some clinical features with typical DA namely; anterior mandibular site predilection, no definite gender predilection and mixed radiolucent-radioopaque radiographic presentation. **Table 1** summarizes the clinical findings of 25 cases of HA reported in the scientific literature.

There have been suggestions that the term "hybrid" if taken literally, might overstate the significance of finding a DA in combination with islands of solid multicystic ameloblastoma, since the term "hybrid tumour" denotes 2 or more disparate and well established tumours that exhibit obvious differentiation. A suggestion that 'hybrid tumour' be considered a "collision tumour" was therefore proposed. [15,23]. Collision tumours are considered to be 2 or more tumours that arise from independent topographic sites. It is possible that the conventional and DA variants of the HA develop simultaneously thereby favoring the collision concept.

4. CONCLUSION

Although the clinicopathological and biological characteristics of HA are yet to be precisely established, it is generally accepted that HA is a variant of the solid mul-

Table 1. Clinico pathologic features of 25 complied cases of Hybrid Ameloblastoma.

Author & Year	Number of cases	Age/Sex	Location	Radiologic Features
Waldron & El Mofty, 1987	5	25 - 82yrs/2-Male	4-Mandible, Posterior	NA
		3-Female	1-NA	NA
Higuchi *et al.*., 1991	2	58yrs/Male	Mandible, Anterior to Posterior	Soap Bubble & Multicystic
		70yrs/Male	Mandible, Posterior	Multicystic
Philipsen *et al.*, 1992	1	55yrs/Male	Mandible, Anterior canine to molar region	Multilocular RL with floccular radiopacities. Root resorption
Ashman *et al.*, 1993	1	53yrs/Male	Mandible, Anterior	Well circumscribed mixed RL & RO
Takata *et al.*, 1999	1	48yrs/Male	Mandible, Lateral Incisor to 1st Molar	Honey comb appearance in anterior region
Wakoh *et al.*, 2002	1	35yrs/Female	Mandible, Canine-premolar region	Unicystic RL in molar region Mixed RL & RO with adjacent cystic radiolucent area
Hirota *et al.*, 2005	1	17yrs/Female	Maxilla, Canine & premolar regions	Mixed RL & RO with well defined borders
Santos *et al.*, 2006	1	36yrs/Male	Mandible, Anterior-premolar region	Ill defined RL
Desai *et al.*, 2006	1	32yrs/Male	Mandible, Posterior	Well defined unilocular RL
Sivapathasundaram *et al.*, 2009	2	31yrs/Female	Mandible, Anterior-premolar	Mixed RL & RO with poorly defined borders
		40yrs/ Male	Maxilla, Anterior-molar	Mixed RL & RO with ill defined borders
Yazdi *et al.*, 2009	1	48yrs/Female	Mandible, Anterior	Mixed RL & RO with ill defined borders
Gade *et al.*, 2010	1	35yrs/Female	Maxilla, Anterior	Mixed RL with radioopaque specks
Gupta *et al.*, 2011	1	35yrs/Female	Mandible, Anterior	Ill defined hazy RL with flecks of RO
Vardhan *et al.*, 2011	1	29yrs/Female	Mandible, Anterior-right body	Well defined irregular RL
Lawal *et al.*, 2011	2	50yrs/Female	Mandible, Anterior	Multilocular RL
		29yrs/Male	Mandible, Anterior	Multilocular RL
Acharya *et al.*, 2011	1	50yrs/Male	NA	NA
Angadi *et al.*, 2011	1	64yrs/Female	Maxilla, Anterior-molar	Ill defined hazy RL & RO, root resorption
Present case	1	50yrs/Female	Mandible, Anterior-molar	Mixed RL & RO

NA—Not Available; M—Male; F— Female, RL—Radiolucency, RO—Radiopacity.

ticystic ameloblastoma. Besides the histological details that combine both conventional ameloblastoma and DA, our present case of HA bears similar clinical and radiological features as well as similar treatment modalities to that of the conventional solid multicystic ameloblastoma. We therefore further add a case of this rare form of ameloblastoma to scientific literature. The frequency of occurrence of HA in our series however appears very low. In the light of our new experience, it would be necessary to revisit our archives with the possibility that we might be able to bring out more cases of HA.

REFERENCES

[1] Barnes, L, Eveson, J.W., Reichart, P. and Sidransky, D. (2005) World Health Organization classification of tumours pathology and genetics of head and neck tumours. IARC Press, Lyon, 283-328.

[2] Dandriyal, R., Gupta, A., Pant, S. and Baweja, H. H. (2011) Surgical management of ameloblastoma: Conservative or radical approach. *National Journal of Maxillofacial Surgery*, **2**, 22-27. http://dx.doi.org/10.4103/0975-5950.85849

[3] Gnepp, D.R. (2009). Diagnostic surgical sathology of the head and neck. 2nd Edition, Saunders, Philadelphia, 785-838.

[4] Gardner, D.G. (1996) Some current concepts on the pathology of ameloblastomas. *Oral Surgery*, *Oral Medicine*, *Oral Pathology*, *Oral Radiology* & *Endodontics*, **82**, 660-669. http://dx.doi.org/10.1016/S1079-2104(96)80441-0

[5] Gomes, C.C., Duarte, A.P., Diniz, M.G. and Gomez, R.S. (2010) Review article: Current concepts of ameloblastoma pathogenesis. *Journal of Oral Pathology & Medicine*, **39**, 585-591. http://dx.doi.org/10.1111/j.1600-0714.2010.00908.x

[6] Buchner, A. and Carpenter, W.M. (2006) Relative frequency of central odontogenic tumors: A study of 1,088 cases from Northern California and comparison to studies from other parts of the world. *Journal of Oral and Maxillofacial Surgery*, **64**, 1343-1352. http://dx.doi.org/10.1016/j.joms.2006.05.019

[7] Lu, Y., Xuan, M., Takata, T., Wang, C., He, Z., Zhou, Z., Mock, D., *et al.* (1998) Odontogenic tumors: A demographic study of 759 cases in a Chinese population. *Oral Surgery*, *Oral Medicine*, *Oral Pathology*, **86**, 707-714. http://dx.doi.org/10.1016/S1079-2104(98)90208-6

[8] Adebayo, E.T., Ajike, S.O. and Adekeye, E.O. (2005). A review of 318 odontogenic tumors in Kaduna, Nigeria. *Journal of Oral and Maxillofacial Surgery*, **63**, 811-819. http://dx.doi.org/10.1016/j.joms.2004.03.022

[9] Adebiyi, K.E., Ugboko, V.I., Omoniyi-Esan, G.O., Ndukwe, K.C. and Oginni, F.O. (2006) Clinicopathological analysis of histological variants of ameloblastoma in a suburban Nigerian population. *Head & Face Medicine*, **2**, 42-46. http://dx.doi.org/10.1186/1746-160X-2-42

[10] Li., W.L., Liu., F.Y., Xu., Z.F., Huang, S.H., Zhu., W., and Sun, C.F. (2012) Treatment of ameloblastoma in children and adolescents. *Journal of Hard Tissue Biology*, **21**, 121-126.

[11] Ladehinde, A.L., Ogunlewe, M.O., Bamgbose, B.O., Lanre, W.L., Ajayi, O.F., Arotiba, G.T. and Akinwande, J.A. (2006) Ameloblastoma: Analysis of 207 cases in a Nigerian teaching hospital. *Quintessence International*, **37**, 69-74.

[12] Hertog, D., Bloemena, E., Aartman, I.H.A., van der Waal, I. (2012) Histopathology of ameloblastoma of the jaws; Some critical observations based on a 40 years single institution experience. *Medicina Oral*, *Patología Oral y Cirugía Bucal*, **17**, e76-82. http://dx.doi.org/10.4317/medoral.18006

[13] Harsha, B.G., Ingle, E., Dornadula, K., Saraswathi, G. (2011) Hybrid lesion of desmoplastic and conventional ameloblastoma—A case report. *Journal of International Oral Health*, **3**, 43-47.

[14] Waldron, C.A., EL-Mofty, S.K. (1987) A histopathologic study of 116 ameloblastomas with special reference to the desmoplastic variant. *Oral Surgery*, *Oral Medicine*, *Oral Pathology*, **63**, 441-451. http://dx.doi.org/10.1016/0030-4220(87)90257-X

[15] Philipsen, H.P., Reichart, P.A. and Takata, T. (2001) Desmoplastic ameloblastoma (including "hybrid" lesion of ameloblastoma). Biologic profile based on 100 cases from the literature and own files. *Oral Oncology*, **37**, 455-460. http://dx.doi.org/10.1016/S1368-8375(00)00111-1

[16] Philipsen, H.P., Ormiston, I.W. and Reichart, P.A. (1992) The desmo- and osteoplastic ameloblastoma. Histologic variant or clinicopathologic entity? Case reports. *International Journal of Oral and Maxillofacial Surgery*, **21**, 352-357. http://dx.doi.org/10.1016/S0901-5027(05)80761-1

[17] Gupta, A. and Jindal, C. (2011) Hybrid ameloblastoma: report of a rare case and review of literature. *International Journal of Oral and Maxillofacial Surgery*, **2**, 68-72

[18] Lawal, A.O., Adisa, A.O., Olusanya, A.A. and Adeyemi, B.F. (2011) "Hybrid" ameloblastoma: A report of two cases. *African Journal of Medicine & Medical Sciences*, **40**, 11-13

[19] Effiom, O.A. and Odukoya, O. (2011) Desmoplastic ameloblastoma: Analysis of 17 Nigerian cases. *Oral Surgery*, *Oral Medicine*, *Oral Pathology*, *Oral Radiology & Endodontics*, **111**, e27-31. http://dx.doi.org/10.1016/j.tripleo.2010.06.021

[20] dos Santos, J.N., De Souza, V.F., Azevêdo, R.A., Sarmento, V.A. and Souza, L.B. (2006) "Hybrid" lesion of desmoplastic and conventional ameloblastoma: Immunohistochemical aspects. *Brazilian Journal of Otorhinolaryngology*, **72**, 709-713.

[21] Yazdi, I., Seyedmajidi, M. and Foroughi, R. (2009) Desmoplastic ameloblastoma (a hybrid variant): Report of a case and review of the literature. *Archives of Iranian Medicine*, **12**, 304-308.

[22] Takata, T., Miyauchi, M., Ogawa, I., *et al.* (1999) So called "hybrid" lesion of desmoplastic and conventional ameloblastoma: Report of a case and review of literature. *Pathology International*, **49**, 1014-1018. http://dx.doi.org/10.1046/j.1440-1827.1999.00972.x

[23] Melrose, R.J. (1999) Desmoplastic ameloblastoma. *Pathology Review*, **4**, 21-27

